

Direct Ascending Pharyngeal Artery to Jugular Vein Arteriovenous Fistula

A Case Report

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Summary

A 32-year-old female developed a bruit, determined to arise from a rare direct arteriovenous (AV) fistula from the ascending pharyngeal artery to the internal jugular vein.

The fistula was treated by transarterial silicone balloon occlusion, with occlusion of fistulous flow, ablation of symptoms, and excellent long-term result.

Introduction

Arteriovenous fistulae from the ascending pharyngeal artery to the jugular vein have been described and illustrated primarily as multiple small feeding arteries, frequently with participation of other dural or osseous arterial contribution¹. Successful occlusion with use of particulate embolic agents confirms the nature of the arterial contribution as smaller arteries without direct AV shunting².

Direct AV fistulae from the external carotid artery to the internal jugular vein have been described, without specific details of the external carotid branch(es) involved. Direct AV fistula from the ascending pharyngeal artery to the internal jugular vein is rare. One instance was included among Berenstein's report of direct external carotid artery fistulae, but neither illustrated nor further described³.

Takahashi reported a direct fistula between the ascending pharyngeal artery and sigmoid

sinus that regressed spontaneously⁴. We report a patient with this rare fistula, including 10-year post-treatment followup, and discuss treatment options and considerations for the disease process.

Case Report

A 32-year-old female developed a pulsatile noise in her left ear, while skiing at high altitude, approximately eight months prior to treatment on 4-5-95. The noise had increased during a recent pregnancy.

Noise and headaches also increased following a recent ski trip. There was no past history of trauma, infection, or hypercoagulability. MR imaging of the skull base region was suboptimal due to artifact from metal teeth braces, and the fistulous connection point could not be precisely localized in relation to bony skull base anatomy.

Cervicocerebral angiography demonstrated a direct arteriovenous fistula between the 3.5-mm ascending pharyngeal artery (likely a branch of the neuromeningeal trunk) and the internal jugular vein near the occipital condyle (Figure 1). On left internal carotid artery (ICA) injection, venous effluence from the left sigmoid sinus was sluggish, but antegrade. A nondetachable balloon was inflated in the distal ascending pharyngeal artery, followed by injection into the common carotid artery, and no additional feeding arteries were identified. Due



Figure 1 Lateral common carotid arteriogram demonstrates the enlarged ascending pharyngeal artery, of approximately the same diameter as the ICA, with direct fistulous connection to the jugular bulb, and venous flow to the internal jugular vein. Note the proximity of the tortuous ascending pharyngeal a. segment to the ICA at the skull base. The segment was overlapped by metal braces in the standard AP view.



Figure 2 Two detachable balloons are in place just proximal to the fistula. The ascending pharyngeal artery–internal jugular vein connection appears to be just at the occipital condyle, also overlapping the lateral mass of C1, on lateral view.

to proximity of the tortuous distal ascending pharyngeal artery to the ICA and skull base bony and muscular attachments, detachable balloons, rather than platinum coils, were chosen as the embolic agent. The fistula was totally occluded with two 0.5 cc. detachable silicone balloons, inflated to 0.2 and 0.15 cc respectively, with metrizamide 200 mg%. No additional or residual fistulous flow was demonstrated from collateral sources. The bruit disappeared. Followup xrays at four weeks, six months, and twelve months demonstrated no change in balloon position or volume.

The patient had a subsequent uneventful vaginal delivery. Follow-up MRI and MRA ten

years later, performed for symptoms of orbicularis oculi spasm and neck aching, demonstrated balloon deflation, a small ascending pharyngeal artery with no evidence of fistula, and no other cervical/intracranial abnormality.

Discussion

Direct AV fistula of external carotid artery branches are relatively uncommon entities, and typically present little immediate risk to patients. Direct fistulous connections from external carotid artery branches to major intracranial-draining veins might cause some risk of generalized or local intracranial hypertension⁶.

Other treatment options for the AV fistula described might include continued observation, n-butyl cyanoacrylate (nbca) adhesive use, coil placement, or direct surgical ligation of the ascending pharyngeal artery. The progressive increase in symptoms, with development of an ascending pharyngeal artery approaching the size of the ICA recommended treatment.

The direct nature of the fistula with the risk of venous passage of liquid acrylic precluded nbca's safe, predictable use. Decision to recommend detachable balloon treatment, rather than pushable-coil treatment, was based on several technical and anatomic factors, including:

- 1) detachable coils were not yet approved for use for this purpose at the time of this patient's management;

- 2) pushable coils were believed to offer a less focal, short-segment complete occlusion;

- 3) soft, compressible nature of the balloon-embolic agent;

- 4) ability to puncture balloons at the skull base for deflation, if necessary;

- 5) temporary nature of balloon inflation and mass effect, expected to deflate over several years;

- 6) unquantifiable risk of the long-term effects of an elongated, compressed coil mass on the ICA at the skull base, particularly with neck rotation or flexion. The relative proximity of the neuromeningeal branch of the ascending pharyngeal artery at the skull base to the internal carotid artery, styloid process, muscle attachments, and lower cranial nerves raised some question of long-term effects of a substantive coil mass (a length of pushable coils of at least 2-3 cm. and 3.5 mm width) to these adjacent structures. Intermittent ICA occlusion has been reported as a result of local compression by adjacent structures;

- 7) unsuccessful balloon treatment would not preclude subsequent treatment by other transarterial or transvenous methods.

Both transarterial and transvenous approaches were considered for treatment. The single arterial approach was chosen.



Figure 3 Post-detachment common carotid arteriogram confirms fistula occlusion with no immediate additional flow beyond the occlusion.

Detachable balloons are no longer available for purchase in the U.S., having been voluntarily removed from the market by the manufacturer. Plans for their return are currently under consideration through a new vendor. Coil treatment has supplanted balloon use following balloon unavailability. Currently, new developments in coil technology, such as hydrocoils, might be expected to create an effective short-segment occlusion, more akin to balloons^{8,9}.

However, detachable balloons remain unique embolic agents, with theoretical advantages and physical characteristics favoring their use under a number of circumstances, such as in the current case report.

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EDITORIAL COMMENT

The authors report a very interesting case of direct arteriovenous fistula that occurred spontaneously in a young female. Beside this 10 year follow up, with the silicone detachable balloon, this case raises some classic issues that one should always have in mind when confronted to such diseases:

- *spontaneous high flow fistula involving the skull base and neck vessels should always suggest collagen disease like Ehlers Danlos. Careful search for familial history of uterine rupture and other organ rupture should be carefully done;*
- *if these lesions are not related to an arterial rupture, they must be included in some sort of arteriovenous malformations revealed or symptomatic at later age. They therefore correspond to what has been described as para-chordal type of lesion which includes parasacral, lumbar, intercostal, vertebro-vertebral, occipito-vertebral, ascending pharyngeal to jugular and internal maxillary AVFs;*
- *clinical manifestations are always related to venous drainage. Some potential cranio-petal drainage should prompt endovascular treatment in order to avoid hypertensive manifestations intra cranially;*
- *treatment can be achieved with multiple modalities, all depending of the presence of a small chamber between the arterial vessel and the draining vein as well as the patency of the anastomoses.*

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